

CASE REPORT

Endoscopic Diagnosis and Management of Iatrogenic Cervical Esophageal Perforation in Extremely Premature Infants

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Blind oro-(naso)-pharyngeal suction and feeding catheter intubation are very common practices in pediatric critical care. However, these simple procedures may produce unexpected complications in friable tiny patients. We encountered 3 extremely premature infants in whom cervical esophageal perforation and further submucosal excavation were caused by traumatic catheter injury and subsequently led to catastrophes. These episodes of iatrogenic trauma were all successfully diagnosed early, documented and managed with the aid of ultrathin flexible endoscopy. [*J Chin Med Assoc* 2007;70(4):171–175]

Key Words: endoscopy, iatrogenic esophageal perforation

Introduction

Catheter interventions in the upper aerodigestive tract, such as the placement of orogastric catheter (OGC), nasogastric catheter (NGC) and the procedure of oro-(naso)-pharyngeal suction are common medical practices. They are often and more aggressively performed on critically ill patients in the intensive care unit (ICU), and rarely result in complications. However, even these simple and well-recognized procedures may occasionally be implicated in hypopharynx or esophagus perforation, especially in a very special group of patients, extremely premature infants who are born with unusually friable tissues, and whose survival can be prolonged with the advent of modern medical facilities but concurrently may have more iatrogenic problems. Both the neonatologist and pediatric surgeon often play a central role in early and correct recognition as well as in offering appropriate management, which are essential to minimize the associated morbidity or even mortality.^{1–6}

Flexible endoscopy (FE) can directly facilitate the discovery of lesion sites^{7,8} which in the past might have escaped detection. When such lesions are found, FE can also demonstrate how the injuries occurred and what

further management should be offered. We encountered 3 extremely premature infants in the past 2 years in whom unexpected cervical esophageal perforations and further deep tissue excavations caused by traumatic suction catheter (SC) and feeding catheter (FC) insertions subsequently resulted in catastrophes. These unusual and frequently unrecognized iatrogenic traumas were demonstrated and appropriately managed by direct ultrathin FE. To the best of our knowledge, this FE application has not been reported in such tiny infants before.

Case Reports

Case 1

A female premature infant of 25 weeks gestational age (GA) and birth weight (BW) 710 g was admitted to the neonatal ICU. She was successfully weaned from oro-tracheal intubation with mechanical ventilation (MV) to nasal prongs with continuous positive airway pressure (CPAP) on the 5th day of life (DOL). Frequent oro-(naso)-pharyngeal suctions with 6-Fr polyvinylchloride (PVC) SC were performed to ensure upper airway

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patency, although some bloody secretion had been noted. In the following days, bolus formula feeding (1 mL/q3h) was given via a 5-Fr PVC OGC, which was changed routinely twice weekly. She gradually developed abdominal distension, which was assumed to be due to CPAP inflation.⁹ A radiograph film on the 14th DOL showed distended bowel loops and a normal OGC position.

On the 15th DOL, due to progressive apnea, desaturation, severe distension, and brown discoloration of the abdomen, the patient was nasotracheally intubated with MV support. A 5-Fr FC was inserted smoothly by an experienced nurse via the other nostril. A radiograph was taken, and at this time, the NGC tip was shown to be abnormally located in the liver field and surrounded by a round density (Figure 1), which led to a high index of suspicion that the NGC was malpositioned and had been wrongly fed.

Immediately, an FE (3.0 mm OD) was performed via the nose, with the erroneous NGC still in place, to search for the exact problem. The esophagus and stomach were initially shown to be normal, but no catheter existed in the lumens. Then, the FE was slowly withdrawn. At the right corner of the esophageal orifice, the NGC was found to be embedded in the mucosa through a 0.7-cm linear laceration wound. Obviously, the NGC had penetrated from this wound to the outside of the alimentary tract. The distance from the



Figure 1. Radiograph shows distended bowel loops. The distal portion of the feeding catheter (arrows) is noted in the liver field and surrounded by radio-opaque shadow.

nostril to the perforation site was measured by the FE to be 6.3 cm. Under direct FE vision, the NGC was removed and a new 5-Fr FC was again nasally inserted and manipulated to avoid reentry into the laceration site. The patient was then conservatively managed with gavage feeding, no deep (no more than 5.5 cm) pharyngeal suction, and no change of NGC. Intravenous broad-spectrum antibiotics were given for a total of 10 days. The following hospital course was smooth, and she was discharged on the 105th DOL in wonderful condition.

Case 2

A male premature infant of BW 1,200 g was successfully extubated from MV to nasal prongs on the 3rd DOL. A 5-Fr PVC OGC was placed to start gastric feeding. Regular oro-(naso)-pharyngeal suction was performed with 6-Fr PVC SC by experienced nurses to keep the airway clear. The SC had met with some resistance around the pharynx region and there were blood-tinged secretions during the suction, but this did not cause much concern. On the 5th DOL, moderate blood came from the OGC. It was initially suspected to be stress associated, and intravenous cimetidine was given, but there was no significant improvement. On the 7th DOL, massive blood flow was seen from the nose and the mouth, and FE was therefore performed.

Under FE vision, the esophagus, stomach and tracheobronchial lumens were all noted to be normal. In the hypopharynx wall near the esophagus orifice, a 0.6-cm linear wound, 7.0 cm from the nostril, was found with blood oozing out continuously. A new SC was inserted via the nose, with repetitive back-and-forth motion in the pharynx. It was observed that the catheter tip penetrated through the wound each time and could tunnel 3 cm into the submucosa without any resistance being felt. We did a test: a new 5-Fr FC was inserted via the other nostril through the pharynx. It was found that the FC always traveled through the customary path to the perforation wound. To maintain gavage feeding, we manipulated the FC down to the esophagus and the stomach, and changed the infant's head posture to avoid the FC going into the wound. Broad-spectrum intravenous antibiotics, no deep (<6.5 cm) pharyngeal suction, and no change of the NGC for 7 days were recommended. After that, the patient did wonderfully and was discharged on the 40th DOL.

Case 3

A female premature infant of 26 weeks GA and BW 920 g was admitted to the neonatal ICU. She had nasotracheal intubation and was successfully weaned to nasal prongs on the 4th DOL. She underwent frequent

oro-(naso)-pharyngeal suction with a 6-Fr PVC SC due to copious secretions after the extubation. Ten hours after extubation, massive fresh blood came from the nose and the mouth, which could not be eliminated with local epinephrine application to the nose and the pharynx. Tachypnea (approximately 70/minute) and desaturation developed gradually. Emergency blood transfusion was recommended. Pulmonary hemorrhage was the first impression, but the chest film showed only mild haziness of bilateral lungs. Therefore, tracheal re-intubation was recommended.

With experience from the previous 2 cases, we asked for a FE check before attempting to intubate. The FE (3.0 mm OD) revealed a 1.0-cm vertical linear perforated wound, 6.5 cm from the nostril, over the right corner of the esophageal opening (Figure 2). All the lower esophageal, stomach and tracheobronchial lumens were shown to be normal. In order to clarify and imitate the cause of injury, a 6-Fr SC was nasally inserted with repeated back-and-forth motion. It appeared that the tip of the SC entered through the laceration wound each time and tunneled a false track about 4 cm deep into the submucosa. For further continued gavage feeding, a new NGC was carefully

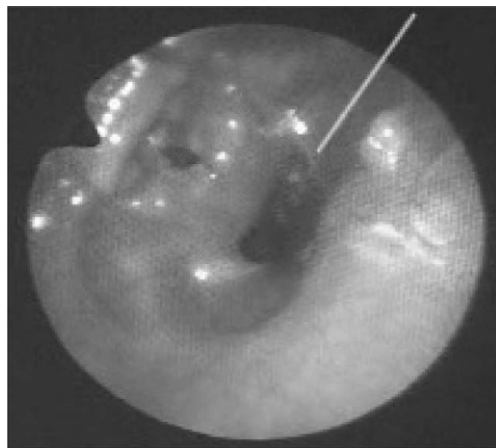


Figure 2. A 1.0-cm vertical mucosa perforation (straight line) is near the right corner of the esophageal opening.

manipulated, under FE vision and with the aid of changing head positions, safely into the stomach. Five-day intravenous antibiotics and prohibition of deep (<6.0 cm) pharyngeal suction were recommended. The infant did wonderfully afterwards.

Discussion

These 3 cases are summarized in Table 1. Iatrogenic perforation of the cervical esophagus is an uncommon but serious complication in critical patients that can easily be overlooked.¹⁻⁶ The time lag of all perforations in our 3 infants were far from tracheal intubation and, therefore, most likely were caused by either suction, gastric intubations, or both. Most perforations associated with catheter manipulation occur during rough, emergent or difficult procedures, which are often performed by relatively inexperienced personnel, and with incorrect use of a big or stiff catheter. This iatrogenic perforation may occur more commonly than is generally thought. If left unrecognized or misdiagnosed, the potential sequelae can be catastrophic, as its clinical propensity mimics oral, nasal, gastric, or even pulmonary injury, to which unstable premature infants are all vulnerable.

Oro-(naso)-pharyngeal suction to maintain upper airway patency is a necessary and beneficial procedure in the care of critically ill patients. This is especially significant in tiny premature infants whose endotracheal tube has just been removed; copious thick secretions coming from the trachea, if pooling and sticking in the upper airway, are prone to blocking these narrow lumens. Patients' weak swallowing and ineffective cough create further difficulties in secretion removal. Therefore, frequent and aggressive suction to clear the upper airway may be inevitable and even highly recommended.

Most suction or gastric catheters available today are made of PVC material and straight at the tip. They may be relatively stiff compared with the premature infant's

Table 1. Characteristics of 3 extremely premature infants with iatrogenic esophageal perforation diagnosed and managed with flexible endoscopy (FE)

Case	Gestational age (wk)	Perforation site	Age (d)	Body weight (g)	Possible causes	Treatment	Prognosis
1	25	Right side of esophageal orifice	15	610	Suction, NG tube	Conservative*	Survival
2	29	Middle hypopharynx	7	1,200	Suction, NG tube	Conservative*	Survival
3	26	Right side of esophageal orifice	4	920	Suction, NG tube	Conservative*	Survival

*Included maintenance of gastric tube feeding, no deep naso-oro-pharyngeal suction and intravenous antibiotics. NG = nasogastric.

mucosa tissue. The path of the human upper airway, from naso-, oro- to hypopharynx, forms almost an S-like shape. When catheters are passed through the nasal or oral tracts, the tips can exert a direct punch force, mainly first on the posterior pharyngeal wall (nasal adenoid, the first curve and the most angulated site) and then on the base of the hypopharynx (the second curve, the narrowed introitus to the esophagus). The pharyngoesophageal junction is especially prone to these punches because the cricopharyngeal muscle easily contracts by local stimulation or injury, becoming more susceptible to trauma and perforation. Extension of the neck, a common manipulation during catheter insertion, can effectively eliminate the first curve, but further increases the possibility of injury at the second curve by exaggerating the tip to produce more punch against the cervical vertebra.¹⁰ Repetitive and excessive striking of the catheter tip might ultimately perforate local mucosa and result in subsequent excavation. This catheter-induced mucosa injury may not be avoided in presence of frail tissue, even if the insertion is done by an experienced clinician. Tiny premature infants, such as our 3 cases here, are particularly vulnerable because of their delicate tissue, the need for frequent catheter (feeding and suctioning) insertion, and inability to immediately respond against this painful injury. Furthermore, premature infants, including all our 3 infants, may frequently receive steroid therapy to facilitate their pulmonary function.^{11–13} Steroids can increase tissue fragility through their adverse effects on collagen formation and have been identified as a possible predisposing factor for perforation.^{14,15}

In case 1, the indwelled FC perforated and tunneled through the pharyngeal submucosa, prevertebral space, and finally reached the right-side retroperitoneum. There was no feeling of significant resistance either during the insertion or the gavage feeding in that deep tissue. This evidence suggests that neonate clinicians and nurses should be more cautious in the use of all instruments that may reach or pass through the premature infant's pharynx and esophagus.

Initial symptoms and signs, including bloody aspirate from oral or nasal cavity, pharynx, catheter resistance and coffee ground gastric residual, which lead to the suspicion that pharyngoesophageal perforation may have occurred, are not specific. If the perforation is not promptly and accurately diagnosed in time, these premature infants may receive unnecessary and/or even hazardous management such as tracheal intubation, positive pressure ventilation, intrapulmonary medications to stop bleeding, antacids, blood transfusion, prohibition of gastric feeding, and prolonged parenteral nutrition, which may cause further complications.

Traditionally, confirmation of a suspected esophageal perforation mainly relies on radiographic images. In some instances, plain neck or chest radiographs may reveal ectopic tube location or abnormal shadow along the catheter pathway. However, plain films may appear normal in 12¹⁶ to 33%¹⁷ of all these cases. A contrast swallow study may disclose the perforation site, a false tract,¹⁸ or wrong localization of the contrast in more detail, but it still has a 10% false-negative rate.^{19,20} Computed tomography to find the extraluminal air is one of the most useful findings for making diagnosis; however it is expensive and may not be suitable for tiny lesions, small catheter and small infants.

FE, as demonstrated in our 3 cases, may be more convenient and accurate for making diagnosis. FE can readily be performed at bedside, therefore avoiding critical delay, transfer to radiology facility, contrast use, and radiation exposure. It is also capable of examining the whole aerodigestive tract, precisely locating the injury site, and directly visualizing how the catheter passes through the mucosa and has made a tunnel. The distance from the nostrils (or lip) to the lesion site can be measured as the reference for further suction depth. In addition, for continuous alimentary nutrition, a new FC can be correctly positioned under FE guidance, to avoid the customary path that would lead to reentry into the original trauma site. Therefore, the infant can be spared from parenteral nutrition, gastrostomy feeding, radiation, and fluoroscopic insertion catheter. In small infants, we perform FE with "a quick look" method: only with topical spray of xylocaine through the nose and pharynx, if indicated, without any sedatives. The total time needed for FE insertion to check the upper aerodigestive tract (including nasal tract, pharynx, larynx, trachea, esophagus, and even stomach) is less than 1 minute. In our experience, this procedure is similar to an SC or FC insertion, and can be safely performed in high-risk infants.^{21,22}

A consensus concerning the management of pharyngoesophageal perforation has developed in recent years. Broad-spectrum antibiotics and immediate exploration with closure and drainage are indicated in the majority of patients, with the exception of neonates.^{1,23,24} Conservative management is still the mainstream treatment for small infants.^{1,4,5,25} The self-recovery courses in our 3 cases suggest that conservative management with FE-guided insertion of FC, maintaining gavage feeding, delicate non-deep suction, and appropriate antibiotics could be successful.

In conclusion, caution should be exercised when performing aerodigestive tract suction, intubation, or using other instrumentation, especially in premature infants. Even with unobvious traumatic evidence,

hypopharyngeal or cervical esophageal perforation should always be suspected in unexplained nasal or oropharyngeal bleeding. Once the perforation is noted, conservative management without deep suction is recommended. FE may be performed in suspected infants prior to other invasive studies, and also for the correct placement of the FC. FE is convenient and worthwhile to quickly make an accurate diagnosis and facilitate clinical management of this iatrogenic injury.

References

- Johnson DE, Foker J, Munson DP, Nelson A, Athinarayanan P, Thompson TR. Management of esophageal and pharyngeal perforation in the newborn infant. *Pediatrics* 1982;70:592-6.
- Cairns PA, McClure BG, Halliday HL, Reid MM. Unusual site for oesophageal perforation in an extremely low birth weight infant. *Eur J Pediatr* 1999;158:152-3.
- Kras JF, Marchmont-Robison H. Pharyngeal perforation during intubation in a patient with Crohn's disease. *J Oral Maxillofac Surg* 1989;47:405-7.
- Panieri E, Millar AJW, Rode H, Brown RA, Cywes S. Iatrogenic esophageal perforation in children: patterns of injury, presentation, management, and outcome. *J Pediatr Surg* 1996;31:890-5.
- Engum SA, Grosfeld JL, West KW, Rescorla FJ, Scherer LRT, Vaughan WG. Improved survival in children with esophageal perforation. *Arch Surg* 1996;131:604-11.
- Lind LJ, Wallace DH. Submucosal passage of a nasogastric tube complicating attempted intubation during anesthesia. *Anesthesiology* 1978;49:145-7.
- Yang JF, Soong WJ, Jeng MJ, Chen SJ, Lee YS, Tsao PC, Hwang B, et al. Esophageal atresia with tracheoesophageal fistula: ten years of experience in an institute. *J Chin Med Assoc* 2006;69:317-21.
- Huang IF, Wu TC, Wang KS, Hwang B, Hsieh KS. Upper gastrointestinal endoscopy in children with upper gastrointestinal bleeding. *J Chin Med Assoc* 2003;66:271-5.
- Jaile JC, Levin T, Wung JT, Abramson SJ, Ruzal-Shapiro C, Berdon WE. Benign gaseous distension of the bowel in premature infants treated with nasal continuous airway pressure: a study of contributing factors. *AJR Am J Roentgenol* 1992;158:125-7.
- Girdany BR, Sieber WK, Osman MZ. Traumatic pseudodiverticula of the pharynx in newborn infants. *N Engl J Med* 1969;280:237-40.
- Durand M, Sardesai S, McEvoy C. Effect of early dexamethasone therapy on pulmonary mechanics and chronic lung disease in very low birth weight infants: a randomized controlled trial. *Pediatrics* 1995;95:584-90.
- Yeh TF, Torre JA, Rastogi A, Anyebuno MA, Pildes RS. Early postnatal dexamethasone therapy in premature infants with severe respiratory distress syndrome: a double-blind, controlled study. *J Pediatr* 1990;117:273-82.
- Rastogi A, Akintorin SM, Bez ML, Morales P, Pildes RS. A controlled trial of dexamethasone to prevent bronchopulmonary dysplasia in surfactant-treated infants. *Pediatrics* 1996;98:204-10.
- Baxter JD, Forsham PH. Tissue effects of glucocorticoids. *AJR Am J Med* 1972;53:573.
- Fauci AS. Glucocorticosteroid therapy. In: Wyngaarden JB, Smith LH, eds. *Cecil's Textbook of Medicine*, Vol. 1, 2nd edition. Philadelphia: WB Saunders, 1985:111-6.
- Han SY, McElvein RB, Aldrete JS, Tishler JM. Perforation of the esophagus: correlation of site and cause with plain film findings. *AJR Am J Roentgenol* 1985;145:537-40.
- Wychulis AR, Fontana RS, Payne WS. Instrumental perforation of the esophagus. *Chest* 1969;55:184-9.
- Jones WG, Ginsberg RJ. Esophageal perforation: a continuing challenge. *Ann Thorac Surg* 1992;53:534-43.
- Bladergroen MR, Lowe JE, Postlethwait RW. Diagnosis and recommended management of esophageal perforation and rupture. *Ann Thorac Surg* 1986;42:235-9.
- Sarr HG, Pemberton JH, Payne WS. Management of instrumental perforations of the esophagus. *J Thorac Cardiovasc Surg* 1982;84:211-8.
- Soong WJ, Jeng MJ, Hwang B. The application of a modified mini-flexible-fiberoptic-endoscopy in pediatric practice. *J Chin Med Assoc* 1995;56:338-44.
- Lee YS, Soong WJ. Flexible endoscopy of aerodigestive tract in small infants. *Pediatr Int* 2003;45:530-3.
- Shockley WW, Tate JL, Stucker FJ. Management of perforation of the hypopharynx and cervical esophagus. *Laryngoscope* 1985;95:939-41.
- Levine PA. Hypopharyngeal perforation: an untoward complication of endotracheal intubation. *Arch Otolaryngol* 1980;106:578-80.
- Dolgin SR, Kumar NR, Wykoff TW. Conservative medical management of pharyngeal perforation. *Ann Otol Rhinol Laryngol* 1992;101:209-15.